CASE REPORT

Ankyloblepharon filiforme adnatum

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Two cases of a rare congenital eyelid abnormality, ankyloblepharon filiforme adnatum, are presented, and novel aspects of treatment, associations and classification are discussed.

A baby was born at 34 weeks' gestational age and weighed 1250 g. There was a history of maternal smoking and methadone use, up to 5 days before delivery. The baby had symmetrical intrauterine growth retardation. Patent ductus arteriosus and a ventricular septal defect were diagnosed by echocardiography. Nasal continuous airway pressure ventilation was given for respiratory distress syndrome. Dysmorphic features were evident: hypertelorism, a small mid-face, clinodactyly and a single band of tissue between right upper and lower eyelids (fig 1). This band was axial, covering the pupil, and prevented full opening of the eyelid. The eyelid was divided by one cut with scissors. No underlying ocular abnormalities were noted. Chromosomal analysis confirmed the diagnosis of Edward's syndrome, and morphine and diuretics were given for 2 days to increase comfort. These were discontinued and the infant died 20 days after birth.

The second baby was born at 40 weeks' gestational age and weighed 3720 g. Pregnancy and delivery were unremarkable. On neonatal examination, no abnormalities were detected except for a skin bridge over both the eyes. The baby's mother and two maternal uncles had a similar congenital abnormality, and were treated at the family home by other relatives. There was no other family history. The baby's skin bridges were each divided by one cut (fig 2). No systemic or ocular sequelae were reported (after five years of follow-up).

DISCUSSION

Ankyloblepharon filiforme adnatum (AFA) is a rare but potentially ambylogenic congenital abnormality of the eyelids, in which single or multiple bands of tissue join the upper and lower eyelid. Treatment is given for neonatal comfort and to exclude the risk to the developing visual system. This report highlights the simplicity in treating AFA. Özyazgan *et al*[1] describe treating AFA under intravenous sedation, but in our patients no sedation or local anaesthetic was necessary. The band of tissue was divided with one cut using scissors, and no bleeding occurred. No patient distress was evident.

AFA is also important, as it can be associated with several disorders, such as trisomy 18 (Edward's syndrome), as in the first case. AFA associated with infantile glaucoma has been reported[2] and systemic associations are seen,[3] including Hay-Wells syndrome (ankyloblepharon-ectodermal dysplasia-clefting syndrome), popliteal-pterygium syndrome (characterised by webbing of the knees), curly hair-ankyloblepharon-nail dysplasia syndrome, and cleft lip and palate.

In the first case, there was a history of maternal methadone use. We are not aware of any evidence suggesting that fetal exposure to methadone may result in neonatal AFA.

Rosenman *et al*[4] proposed a classification of AFA into four groups. Bacal *et al*[5] suggested a fifth group: AFA in association with chromosomal abnormalities, and the first case fits in group 5.

The second case had a family history of AFA, but no other family history of note, such as cleft lip or palate. We suggest that such cases could be included in a new group, group 6. Only one other case has been reported of apparently inherited and isolated AFA that would fit in group 6.[1] The family history of AFA in the second case suggests an autosomal dominant pattern of inheritance. A plausible candidate gene perhaps worthy of study in cases of isolated inherited AFA may be *p63*, given its putative role in development, and the varied consequences of mutations in this gene, including Hay-Wells Syndrome.

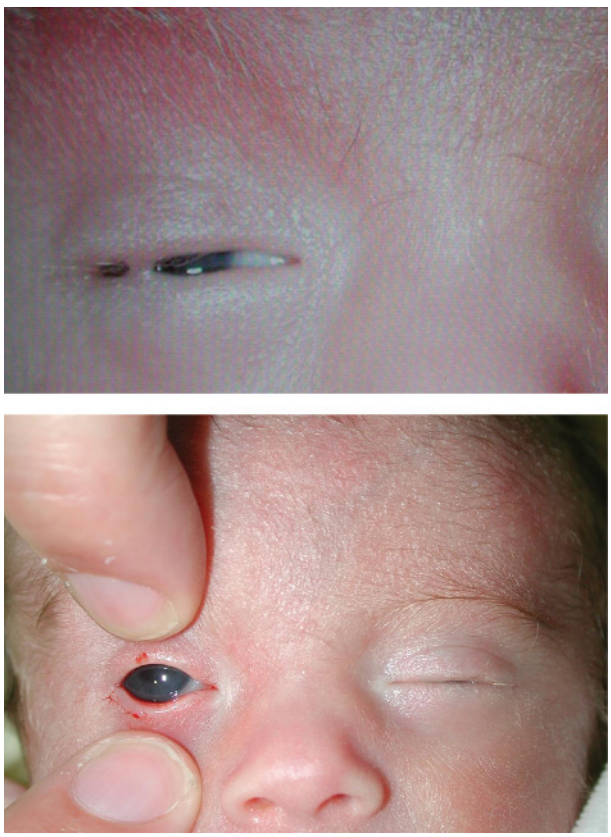

Figure 1 (A) Before treatment. (B) Immediately after treatment (cornea was clear on removing mucus).

Abbreviation: AFA, ankyloblepharon filiforme adnatum


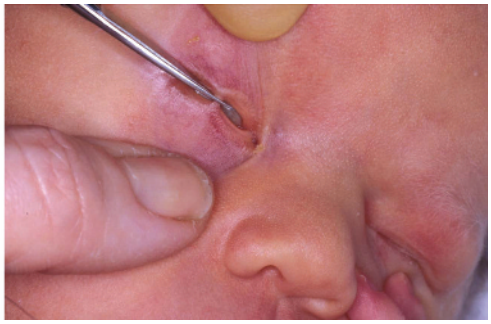

Figure 2 (A) Before treatment (sclera showing due to Bell's Phenomenon). (B) Treatment.

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ARCHIVIST

Gastrostomy feeding

Swallowing difficulties, gastro-oesophageal reflux and pulmonary aspiration are all common among children with neurodevelopmental disabilities. They may result in poor nutrition and growth, lung disease, poor immune function, oesophageal pain, and social and emotional upheaval for child and family. Reviews of the benefits of gastrostomy have been inconclusive, with little attention being paid to psychosocial aspects.

A prospective case series has been reported from the Great Ormond Street Hospital, London, UK (GM Craig and colleagues. *Developmental Medicine and Child Neurology* 2006;**48**:353–60).

In all, 92 families and children were invited to take part in the study and 76 children (44 boys and 32 girls) with a median age of 3 years, 4 months did so. Their diagnoses were cerebral palsy (n = 32), chromosomal or other genetic syndromes (n = 25), slowly progressive degenerative disease (n = 11) and unconfirmed (n = 8). The type of surgery was percutaneous endoscopic gastrostomy in 39 (51%) children, open gastrostomy in 31 (41%) children and laparoscopic gastrostomy in 6 (8%) children. An anti-reflux procedure was performed in 35 (46%) children.

After gastrostomy, a marked catch-up growth in weight for age and mid–upper arm circumference was noticed. Before gastrostomy, 64% of the children had weights of ⩾–2 standard deviation scores (SDS) and 42% of the children had weights of ⩾–3 SDS. After gastrostomy, the corresponding proportions were 54% and 33%. Catch-up growth was achieved by 70% of the children. Dietary assessments for 65 children did not show improved nutritional intakes postoperatively. Marked postoperative improvements in drooling, secretions, vomiting and constipation were noticed. In all, 13 children had at least one major complication of surgery and 61 had at least one minor complication.

Feeding gastrostomy may result in catch-up growth and health gains. The economic and psychosocial aspects, including the parents' views about whether the procedure was worthwhile, are not reported in this paper.